Understanding and identifying key issues with the involvement of clinicians in the development of decision-analytic model structures: A qualitative study

Husbands, Samantha; Jowett, Sue; Barton, Pelham; Coast, Joanna

DOI:
10.1007/s40273-018-0705-7

Document Version
Peer reviewed version

Citation for published version (Harvard):
Husbands, S, Jowett, S, Barton, P & Coast, J 2018, 'Understanding and identifying key issues with the involvement of clinicians in the development of decision-analytic model structures: A qualitative study', *PharmacoEconomics*, pp. 1-10. https://doi.org/10.1007/s40273-018-0705-7

Understanding and identifying key issues with the involvement of clinicians in the development of decision-analytic model structures: A qualitative study

Samantha Husbands[1], Susan Jowett[2], Pelham Barton[3], Joanna Coast[4]

1. Population Health Sciences, Bristol Medical School, University of Bristol, England
2. Health Economics Unit, Institute of Applied Health Research, University of Birmingham, England
3. Health Economics Unit, Institute of Applied Health Research, University of Birmingham, England
4. Population Health Sciences, Bristol Medical School, University of Bristol, England

Correspondence to: Samantha Husbands, Population Health Sciences, University of Bristol, 1-5 Whiteladies Road, Bristol BS8 2RP. Telephone: 01174283121. Email: Samantha.husbands@bristol.ac.uk

Abstract

Introduction: Decision-analytic models play an essential role in informing healthcare resource allocation decisions; however, their value to decision makers will depend on model structures being clinically valid to determine cost-effectiveness recommendations. Clinician involvement can help modellers to develop clinically valid but straightforward structures, however, there is little guidance available on methods for clinician input to model structure. This study aims to provide an in-depth exploration of clinician involvement in structural development, highlighting key issues and generating recommendations to optimise practices.

Methods: A qualitative study was undertaken with a range of modellers and clinicians working in different modelling contexts. In-depth interviews and case studies using observations were carried out to understand how clinicians are involved in model structural development and to identify problems and optimal approaches from informants' perspectives.

Results: Twenty-four interviews and two case studies were undertaken with modellers and modelling teams. Key issues included the number and diversity of clinicians contributing to structural development, potentially impacting the generalisability of structures, and problems with clinician understanding of important information to contribute to model pathways. Modellers and clinicians suggested that clinician training in modelling could enhance structural processes.

Conclusions: Recommendations to optimise current practices include recruiting clinicians from a variety of backgrounds and using discussions between experts to develop valid and generalisable structures. Future research should focus on developing training materials for clinicians and finding ways to help modellers recruit clinicians from different settings.

Key points

- Clinician input to the development of decision-analytic model structures is important to ensure clinical validity; however, little guidance is available on optimal methods.
- Key issues with clinician involvement in structural development include the generalisability of model structures, and clinician understanding of the information that it is important to contribute for the development of model pathways.

- This research recommends that modellers recruit or involve clinicians from a variety of backgrounds to achieve representative model pathways, and consider offering clinicians training in model development to enhance communication and structural processes.

Acknowledgements

Samantha Husbands was primarily responsible for conducting and analysing the qualitative research and for the conception and drafting of the manuscript. Susan Jowett, Pelham Barton and Joanna Coast assisted with the conception of the manuscript and critically reviewed early drafts. Joanna Coast analysed a proportion of the qualitative interviews with modellers. All authors read, edited and approved the final manuscript.

1 Introduction

Decision-analytic models play a key role in informing health technology assessment (HTA) and resource allocation decisions [1, 2]. Models provide a vehicle for economic evaluation and cost-effectiveness analysis, where the focus is on establishing incremental costs and consequences of competing interventions to inform decision-making [3]. An essential aspect of developing a model is ensuring that it is clinically valid and reflects real life, but also that it is straightforward to its users [4]. Model structures can therefore be more simplistic than the 'real world'. The pathways or states contained in model structure should reflect the disease natural history [5-8] but also prioritise events where differences in the costs and outcomes between competing interventions are expected, and events that are likely to impact the model output [9,10]. Consulting with clinicians can help modellers to build straightforward but valid structures and ensure important events are captured. However, little guidance exists on clinician input to structural development, with current guidelines suggesting that clinicians should be consulted but offering limited insight into how this works in practice [8]. A recent synthesis of modelling guidelines ignored clinician involvement in structural development entirely [11].

The benefits of using qualitative research to explore modelling activities have been advocated, with a view to using findings to make improvements to modelling practices [12]. A small number of empirical studies have used qualitative methods to generate recommendations for model-building, looking at clinician involvement in structural development as part of broader research [13-15]. Recommendations from these studies emphasised the importance of using clinical input to translate the disease area/decision problem into model structure, and for structural validation. These papers suggested that clinicians should be involved early in structural development [15] and recommended the input of large numbers of clinicians [8] and clinicians from different backgrounds [13]. Two papers suggested methods for facilitating communication between modellers and clinicians, specifically non-technical terminology and structural diagrams to aid discussions [13, 14]. Although these recommendations are valuable, there has been no focused research to understand current practice and key issues with clinician involvement in structural development.

This paper presents findings from a research study using in-depth qualitative methods to explore clinician involvement in structural development, aiming to highlight good practice and important areas for future research. Model structure is defined here as the representation of the health and economic consequences occurring when patient populations receive particular medical interventions [16].

2 Methods

2.1 Overview

This research used two phases of qualitative study to investigate clinician involvement in structural development from the perspectives of those involved [17]. In-depth interviews were used in the first phase to gather rich accounts of modellers' processes for involving clinical experts [18]. The second phase used case study methods, specifically non-participant observation and semi-structured interviews, to gain comprehensive insight into structural development as it occurred [19]. Whilst observations demonstrated how modelling teams were involving clinicians, interviews captured reflections of informants on methods used. Both phases of the research were used together to generate key findings.

2.2 Sampling

Modellers for in-depth interviews were sampled using purposive sampling, which focuses on the views of those able to provide in-depth knowledge of the topic of interest [19]. Sampling also aimed to capture variation in breadth of modeller experience, from two settings where economic modelling is integral to HTA: UK and Canada [20, 21]. Modellers approached included those working in academia, industry (for consultancies/pharmaceutical companies), and policy (institutes). The seniority of an informant (level of experience) was judged according to whether modellers worked mostly in a managerial capacity (senior), or on hands-on model development (junior). Snowball sampling, where existing informants suggest others to contribute to the research, provided access to non-academic and international informants [22]. Sampling continued until saturation; where no new insights were emerging from continued interviews [23].

Sampling for case studies involved selecting modelling teams to observe throughout a single model's development, with those who could provide potentially different insights to enhance transferability of findings [24]. Sampling focused on teams who had a good reputation in model building and from whom we could make inferences about good modelling practices, which would be helpful to other modellers and settings [25]. Phase one informants were asked to recommend modelling teams who met these criteria, and a sub-sample were approached by the authors.

2.3 Data collection

In-depth interviews were conducted face-to-face [26]. Interviews followed a topic guide (Appendix 1) which was updated as new themes emerged. Open-ended, responsive questioning was adopted [27].

Case study observation was undertaken of all face-to-face meetings and emails, and semi-structured interviews were conducted with modellers and clinicians at various time points throughout structural development. Most interviews were face-to-face, but some with clinicians were over the telephone. A topic guide was used, which included general questions and those specific to context observed.

Meetings were audio-recorded, and notes were taken to report the primary author's perceptions and thoughts [28]. Interviews in both phases were audio-recorded.

2.4 Data analysis

In-depth interviews were analysed by SH using methods of constant comparison, which requires new data to be continually compared with existing data to enhance understanding and develop key themes [29]. Interview transcripts were coded line-by-line and data were organised into themes and assigned a representative code [30]. Analysis was undertaken for batches of interviews and a coding structure was developed and applied to all transcripts, with codes continually updated for new data. Analytic accounts were created for each batch of interviews to compare informants' comments, and later combined. A sub-sample of transcripts were double-coded by JC.

Case study data were analysed using Framework analysis; a matrix based analytic method where individual informants' data (for example excerpts from interviews) are charted and organised into shared themes to synthesise findings [31]. Analysis began with a review of data, and notes taken of important and recurrent themes to generate a thematic framework [31]. This framework was applied to all case study materials and refined as themes emerged. Data were coded by line or passage using a qualitative software package, NVivo10.

Charts were created for each case study to organise excerpts from data collection according to key themes and the method by which they were generated.

In presenting findings, all informants have been assigned identifiers starting M to indicate a modeller and C a clinician; clinical details of modelling activities are anonymised. The findings have been synthesised under four overarching themes which combine findings from both research phases: recruiting clinicians, number and background of clinicians, problems with clinician involvement and enhancing clinician involvement.

3 Results

3.1 Informant characteristics

For phase one, 24 in-depth interviews were undertaken with a range of modellers, with varying experience noted within senior and junior groups (Table 1).

For phase two, two case studies were undertaken. Case Study A (CSA) was within a UK university, with a junior and senior modeller and one clinician (C1) involved in the primary team. C1 was the Chief Investigator (CI) and was a specialist consultant with over ten years' experience working for the National Health Service (NHS). C1 had no previous experience of modelling. The model received ad-hoc input from another modeller and wider group of clinical collaborators and statisticians. Case Study B (CSB) was in a UK policy institute and the modelling team included a junior and senior modeller, and up to eleven clinical experts per meeting. The backgrounds of clinicians recruited to CSB varied in geographical area and clinical role. A subset of three clinicians were sampled for interview (C2-C4). All worked for NHS hospitals and had over ten years' experience as a consultant in their respective specialisms. All also had some previous experience of health economics, with C3 having input to other models and C2 and C4 worked on research projects with a cost-effectiveness element. Other members of the modelling team included information specialists, systematic reviewers and project managers. See Table 2 for case study characteristics. Table 3 contains a summary of the results.

3.2 Recruiting clinicians for structural development

Modellers interviewed in phase one discussed clinician recruitment, with a common scenario for UK academic modellers, and CSA, being that clinicians were CIs or co-applicants on a project requiring modelling expertise. The industry and policy modellers interviewed (including CSB) mostly reported recruiting clinicians to their own model-based projects, citing formal and established methods for doing so (including links through model clients and via public advertisements). However, the remaining academic modellers (UK and Canadian) described informal processes for recruitment, such as cold calling and pursuing links through colleagues. These modellers indicated that they had difficulty engaging clinicians, particularly when relying on clinicians for ad-hoc input:

> M4 (interview, phase 1): *"it's often quite difficult to get a clinician's time… if they have a lot of clinical time with patients it's almost impossible..."*

In terms of a clinicians' role, there was consensus across interviews and case studies that clinician input was required to inform model clinical pathways, and emphasis from some modellers around the importance of clinicians agreeing a final version of structure prior to running the model:

M24 (interview, phase 1): *"I don't do too much on [validation of] structure…if I've got sign off from the clinician…"*

3.3 Number and background of clinicians

Numbers of clinicians involved in structural development varied for interview informants (Table 4), as they discussed involving between zero and twelve, with the most common scenario being two, but a quarter of informants discussing working with one. CSA and CSB involved one and up to eleven clinicians respectively. Numbers differed as those working in industry and policy involved greater numbers than those in academia, potentially due to different strategies for recruitment. Modellers suggested numbers could vary between projects and according to disease complexity and variation in healthcare practices:

M9 (interview, phase 1): *"... [Disease area] is where you have the biggest variation in clinical practice and I think you need bigger numbers because even within a hospital trust you'd see different practices in terms of current care…"*

A third of modellers interviewed discussed the importance of working with 'key' or senior clinicians, implying that it was likely to increase a model's clinical validity and robustness to outside criticism:

M13 (interview, phase 1): *"You want as many leading people in the field as possible because hopefully you will have got all of the opinions out prior to [submission] …you're not going to have some clinician turning up saying something different"*

3.4 Problems with clinician involvement

3.4.1 Clinician numbers

Although a quarter of modellers described instances of involving one clinician in structural development; modellers' general opinion was that this would limit structural generalisability:

M7 (interview, phase 1): *"…you're asking one person to make an assumption… in their experience it may be completely different than someone else's…"*

M12 (interview, phase 1): *"you could have a rogue, which has happened…one clinician view, fine, went to the board and they said 'the way he treats patients is completely different'…"*

Issues with generalisability and clinician numbers were raised in CSA, as C1, being the only clinician involved in the primary team, struggled to decide on a representative care pathway, having worked in centres with different practices:

C1 (observation, CSA): *"…in [Location 1] the consultant would treat the [Test 2] result rather than… the symptoms …but in [Location 2] because we have [Test 1]… if we can find [Symptom 2] we will do [Minor surgery]"*

However, there were a small number of modellers who discussed problems with involving larger groups in structural development, specifically managing and incorporating multiple perspectives:

M17 (interview, phase 1): *"…things quickly get complicated because you end up with eight clinicians who have eight different views… and end up with an over-elaborate model…"*

The issue of generalisability of clinician experience was rarely acknowledged, with only one interview informant (M8) and one case study (CSB) having discussed or observed recruiting clinicians from a range of backgrounds:

> M8 (interview, phase 1): *"… an A and E nurse, a consultant…a surgeon…we try to get a big range of clinicians involved with the pathway…"*

3.4.2 Clinician understanding

Most interview informants reported problems with receiving information from clinicians, as clinicians focused on the experiences of individuals rather than groups of patients when informing structural pathways:

> M11 (interview, phase 1): *"this concept of 'we are massively oversimplifying what happens in this disease and we're assuming every patient is the same', that's quite difficult for clinicians"*

> M14 (interview, phase 1): *"the hard thing with clinicians is getting them to abstract because they see individuals, they don't see a group"*

Many interview informants also commented on the tendency of clinicians to highlight uncommon and unrepresentative patient experiences when suggesting health states/pathways to include:

> M4 (interview, phase 1): *"they'll talk about really rare events…that in their thirty years of experience they've witnessed once, and they'll suggest you put this in…"*

Similar tensions were observed within CSA, as C1 suggested that the experiences of a rare patient group should be incorporated, with the modeller conversely suggesting that this population should be excluded for not representing a common group:

> Observation, CSA:

> C1: *"Include them…in the last six years…I have only seen one a year where they've had [Problem 3] and they've had [Major surgery], they're a small proportion…"*

> M26: *"…we just want to be trundling along as if most people are the general case…although you've said include them I think we mean exclude them, because they're such a minority…"*

However, a contrasting situation was observed in CSB, as clinicians appeared to consider it inappropriate to include a rarer clinical event in the structure due to lack of available evidence. The modeller subsequently implies that this event should be included because it is economically important and may impact the cost-effectiveness analysis:

> Observation, CSB:

> M28: *"Can those with a false positive diagnosis have [Treatment]…?"*

> C4: *"It could happen…."*

> C5: *"…I've certainly heard of a case…"*

> C10: *"I think on the basis of [finding] evidence you can say no…."*

M28: *"…but that box still needs to be explored to compare [Comparator C] against how many people would end up there with [Comparator B] or [Comparator A]"*

Other communication issues observed in the case studies included clinicians' problems with interpreting the research and economic terminology used by modellers:

Observation, CSA:

M25: *"are the utilities, after the surgery…"*

C1: *"…sorry?"*

M25: *"…if the person has [Major surgery], the utility might be different…"*

C1: *"…the, what will be different sorry?"*

M26: *"…the quality of life…"*

3.5 Enhancing clinician involvement: recommendations emerging from the research

3.5.1 Optimum number

Observations from CSB suggested that discussions between multiple clinicians were valuable for identifying common and representative structural pathways, avoiding the generalisability issues observed with one clinician in CSA:

Observation, CSB:

C5: *…[if] the [Comparator A test] is negative and I've got low…clinical suspicion, there's a pathway back out the door…"*

C3: *"…once you've [tested] someone for [Problem A], we are obliged to put them on a pathway…"*

C5: *"… you always refer the patient into…follow-up?!"*

C9: *"…. it varies from clinic to clinic…but people get a second [test] even if they are asymptomatic…"*

C3: *"I think the answer is 'yes, a second [Comparator A test] gets done'…"*

Comments from clinicians in CSB advocated involving larger numbers and a diversity of clinical expertise, with discussions leading to increased confidence in the clinical validity and structural generalisability of model results:

C2 (interview, CSB): *"It's clear from discussions around the table that things are very different across the country and …it's…really important…in terms of validating the outcome…the strength and breadth of experience and geography of clinicians"*

C3 (interview, CSB): *"If there are issues that you haven't considered as a group that are pointed out after [the model has been published] it would undermine credibility…it's important to have these discussions"*

3.5.2 Clinician training

Several modellers suggested offering clinicians training in structural development to enhance efficiency of structural processes, advocating the value of providing clinicians with information about clinically and economically important pathways/states to communicate:

> M24 (interview, phase 1): *"…clinicians who have got training…just totally get it…they know which [health states/pathways] are a big deal and which ones…aren't really going to influence results…"*

> M25 (interview, CSB): *" …it is necessary for [clinicians] to understand…the health economics and mostly what drives the costs and what is important in the clinical effectiveness of the treatment…"*

The idea of training was supported by all clinicians interviewed, as each discussed their own struggles with understanding. Clinicians suggested that guidance from modellers on how models work, modelling terminology and expected contributions to structural development would be beneficial:

> C1 (interview, CSA): *"…we could have a…brief in the beginning to explain health economics, what modelling is, and what they expect from us…"*

> C2 (interview, CSB): *"…a dictionary of terms…a glossary…'this mean[s] this'"*

Clinicians commented on the preferred nature of training, suggesting interactive and distance learning were optimal.

> C3 (interview, CSB): *"…interactive online training or a video…because then it would be something we could do in our own time…"*

Clinicians implied that regular education was important, suggesting that seeing updates of the model as it was developed would enhance understanding. This could help optimise clinical validity through regular checks, and aid clinicians with interpretations of results. A few modellers reflected that showing clinicians structural diagrams during development helped with engagement:

> C4 (interview, CSB): *"…models are such an unfamiliar way of looking at information….I don't [understand] the shape the model is taking…I suspect the finished product [is] going to take some explaining…."*

> M13 (interview, phase 1): *"…when you're going through a model physically they [clinicians] get more involved…"*

However, there was resistance from a small number of modellers, who questioned the practicality of work required to develop and deliver training materials:

> M26 (interview, CSA): *"….you don't want to meet every clinical collaborator and sit down and tell them why [modelling is] needed…"*

4 Discussion

4.1 Key findings

This research has provided detailed insight into clinician involvement in structural development, highlighting key issues around clinician recruitment, numbers and generalisability, and clinician understanding. Information

was collected on numbers and backgrounds of clinicians involved in current processes, demonstrating the average number to be two, but highlighting the experiences of several modellers who had involved only one. This included the observation of a single clinician in Case Study A, who struggled to decide on representative clinical pathways to inform model structure. Only one of the case studies and one modeller interviewed discussed recruiting clinicians from different clinical/geographical backgrounds. These findings raise issues around the generalisability of model structures, particularly when compared to approaches used to elicit evidence parameter values from experts, as these studies recruit/recommend greater numbers and varied samples of clinicians to reflect differences in clinical practices and avoid bias [32, 33]. Further, several modellers in this research discussed being approached for participation by clinicians with existing projects (academic modellers) or using established contacts to recruit experts (industry modellers), with emphasis on speaking to leading and senior clinicians. However, this approach could potentially lead to bias in similar groups of clinicians continually being asked to contribute, and the under representation of experiences of lesser known and junior clinicians.

Modellers reported and were observed having difficulties with retrieving required information from clinicians, as clinicians were unaware of the importance of including events in the structure which are likely to impact cost-effectiveness. Clinicians had problems understanding how model structures were developed and with interpreting the economic and research terminology used, with clinicians and modellers supporting training for clinicians. Although we have not identified any studies advocating clinician training in structural development; its importance has been emphasised in the expert elicitation literature, suggesting that experts will give more confident and accurate answers if they know the purpose and processes behind the tasks they are given [32].

These findings collectively support the value of recommendations to enhance clinician involvement in structural development.

4.2 Strengths and limitations

This research extends insight into clinician involvement in structural development significantly, based on in-depth qualitative study with a broad sample of modellers. Although there were synergies between the findings generated and those of similar studies, including the importance of effective communication and varied clinical perspectives [13-15], issues around generalisability and the importance of clinician training have not previously been emphasised. A comprehensive sampling strategy was employed to include the perspectives of a range of modellers to ensure findings from this research could be generalisable to multiple modelling contexts and relevant to a variety of modellers. The research also includes the perspectives of clinicians on structural development, which have not been investigated previously.

The work was limited by only having resources to conduct two case studies, both in the UK setting. Although the case study research was extensive, further work could benefit from widening the case studies to explore models conducted within industry, in other country settings or with smaller/lesser known modelling teams. It is possible that the work is also less relevant for those working outside of the UK and Canada, where modelling practices may be different. Greater insight may have been permitted from further clinician interviews, as sampling was limited by the availability of clinicians in each case study, and it may be that their views and experiences are not representative of all clinical experts. Nevertheless, in-depth qualitative methods have

allowed detailed and novel insights to emerge, including through observation of communication between modellers and clinicians. Such an extensive investigation of real structural development processes has resulted in recommendations focusing on issues reported and observed to be most pertinent to those developing models [12].

4.3 Recommendations for practice and future research

Findings suggest that a purposeful approach to sampling clinicians for structural development is optimal, aiming to achieve maximum variation in clinicians' backgrounds relevant to the modelling context [34]. Sampling therefore focuses on the diversity of experts' experiences, rather than maximising numbers, aligning with recommendations from expert elicitation studies [32, 33, 35] and acknowledging the recruitment difficulties reported. Modellers should make efforts to sample junior clinicians and those working outside of leading centres and professional links, to avoid correlation in clinicians' views and to account for potentially different perspectives. The practicality of sampling clinicians outside of typical avenues is an important consideration for future research. Discussions between clinicians were observed to be valuable in identifying representative model pathways, and as such modelling processes may benefit from structured methods for gathering and managing multiple clinician perspectives. Possible approaches include qualitative focus groups [12] and the nominal group technique [32], which are both overseen by a moderator to avoid dominance and bias, and encourage individuals to reach consensus through consideration of each other's views.

The development of clinician training is an important area for future work, requiring further research to generate and evaluate 'universal' training content, with emphasis on the issues identified as important for guidance here. However, it is perhaps also worth considering the value of educating modellers in how to optimally present and gather information from clinical experts. Where time is limited, structural development may still benefit from modellers providing clinicians with a short history of decision-analytic modelling, explaining how structural pathways differ from clinical practice and providing regular updates on structural development using diagrams.

It would be interesting to conduct further case studies to explore structural development in models where there are strong clinical advocates for the alternatives; where biases in pathway development may arise. Another important area for empirical research is exploring the methods and approaches modellers are using to elicit and aggregate quantitative evidence parameters from clinicians [35]. More broadly, qualitative research could explore other structural questions, such as how modellers incorporate clinicians' views alongside information from other sources, including literature, other models and data availability.

5 Conclusions

This qualitative study has provided detailed insight into clinician involvement in structural development, highlighting key issues from modeller and clinician perspectives. Recommendations for modellers include recruiting clinicians from diverse backgrounds to encourage generalisable structures, and facilitating discussions between clinicians to generate robust and representative structural pathways. Future research should focus on clinician training to enhance efficiency of structural development processes, and investigate strategies for sampling clinicians outside of typical approaches. Steps to optimise clinician involvement can enhance the clinical validity of model structures and increase confidence in the decisions models inform.

Funding: Financial support for this study was provided in part by a scholarship from the University of Birmingham, and in part by a PhD Scholarship from Universitas 21 for the Canadian element of the work.

Conflict of interest: Samantha Husbands, Susan Jowett, Pelham Barton, and Joanna Coast declare they have no conflict of interest.

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Ethical approval was granted from the Science, Technology, Engineering and Mathematics Ethical Review Committee at the University of Birmingham and the University of British Columbia Behavioural Research Ethics Board (references: ERN_12-1553 and H13-01796).

Informed consent: Informed consent was obtained from all individual participants included in the study.

Data availability: The data generated during the current study are not publicly available due to the highly identifiable nature of the data collected from the interviewees and about the case studies.

References

1. Sculpher MJ, Claxton K, Drummond M, et al. Whither trial-based economic evaluation for health care decision making? J Health Econ. 2006;15(7):677-87.
2. Petrou S, Gray A. Economic evaluation alongside randomised controlled trials: design, conduct, analysis, and reporting. BMJ. 2011;342:d1548.
3. Gray AM, Clarke PM, Wolstenholme JL, Wordsworth S. Applied Methods of Cost-effectiveness Analysis in Health Care. Oxford: Oxford University Press; 2011.
4. Sculpher M, Fenwick E, Claxton K. Assessing Quality in Decision Analytic Cost-Effectiveness Models: A Suggested Framework and Example of Application. Pharmacoeconomics. 2000;17:461–477.
5. Drummond M, Sculpher M, Torrance G, O'Brien B, Stoddart G. Methods for the Economic Evaluation of Health Care Programmes. 4th ed. Oxford University Press, Oxford; 2015.
6. Philips Z, Bojke L, Sculpher M, et al. Good practice guidelines for decision-analytic modelling in health technology assessment. Pharmacoecon. 2006;24(4):355-71.
7. Sun X, Faunce T. Decision-analytical modelling in health-care economic evaluations. The Eur J Health Econ. 2008;9(4):313-23.
8. Roberts M, Russell LB, Paltiel AD, et al. Conceptualizing a model: A report of the ISPOR-SMDM modeling good research practices task force–2. Med Decis Making. 2012;32(5):678-89.
9. McCabe C, Dixon S. Testing the Validity of Cost-Effectiveness Models. Pharmacoeconomics. 2000;17: 501–513.
10. Soto J. Health Economic Evaluations Using Decision Analytic Modeling: Principles and Practices - Utilization of a Checklist to Their Development and Appraisal. Int. J. Technol. Assess. Health Care. 2000;18,:94–111.
11. Peñaloza Ramos MC, Barton P, Jowett S, et al. A systematic review of research guidelines in decision-analytic modeling. Value Health. 2015;18(4):512-29.
12. Husbands S, Jowett S, Barton P, et al. How Qualitative Methods Can be Used to Inform Model Development. PharmacoEcon. 2017;35(6):607-12.
13. Chilcott J, Tappenden P, Rawdin A, et al. Avoiding and identifying errors in health technology assessment models: qualitative study and methodological. Health Technol Assess. 2010;14(25).
14. Kaltenthaler E, Essat M, Tappenden P, et al. Identification and review of cost-effectiveness model parameters: a qualitative study. Int J Technol Assess Health Care. 2014;30(3):333-40.
15. Squires H, Chilcott J, Akehurst R, et al. A framework for developing the structure of public health economic models. Value Health. 2016;19(5):588-601.
16. Kuntz KM, Weinstein MC. Modelling in economic evaluation. In: Drummond M, McGuire A, editors. Economic Evaluation in Health Care: Merging Theory with Practice. New York: Oxford University Press; 2001. p. 141-171.
17. Flick U, von Kardorff E, Steinke I. What is Qualitative Research? An Introduction to the Field. In: Flick U, von Kardorff E, Steinke I, editors. A Companion to Qualitative Research. London: Sage Publications; 2004. p. 3-11.
18. Green J, Thorogood N. Qualitative methods for health research: 2nd ed. London: Sage Publications; 2013.
19. Patton MQ. Qualitative Research & Evaluation Methods. 3rd ed. California: Sage Publications; 2002.

20. Firestone WA. Alternative Arguments for Generalizing From Data as Applied to Qualitative Research. Educ Res. 1993;22(4):16-23.
21. Mays N, Pope C. Qualitative research in health care: Assessing quality in qualitative research. BMJ. 2000;320(7226):50.
22. Kuper A, Lingard L, Levinson W. Critically appraising qualitative research. BMJ. 2008;337:a1035.
23. Bowen GA. Naturalistic inquiry and the saturation concept: a research note. Qual Res. 2008;8(1):137-52.
24. Polit DF, Beck CT. Generalization in quantitative and qualitative research: Myths and strategies. Int J Nurs Stud. 2010;47(11):1451-8.
25. Flyvbjerg B. Five Misunderstandings About Case-study Research. Qual Inq. 2006;12(2):219-45.
26. Legard R, Keegan J, Ward K. In-depth Interviews. In: Richie J, Lewis J, editors. Qualitative Research Practice: A Guide for Social Science Students and Researchers. London: Sage Publications; 2003. p. 138-69.
27. Rubin HJ, Rubin IS. Qualitative Interviewing: The Art of Hearing Data. 2nd ed. Thousand Oaks: Sage Publications; 2005.
28. Miles MB, Huberman AM. Drawing Valid Meaning from Qualitative Data: Toward a Shared Craft. Edu Res. 1984;13(5):20-30.
29. Glaser BG. The Constant Comparative Method of Qualitative Analysis. Soc Probl. 1965;12(4):436-45.
30. Strauss A, Corbin J. Basics of Qualitative Research: Grounded Theory Procedures and Techniques. Newbury Park: Sage Publications; 1990.
31. Ritchie J, Spencer L, O'Connor W. Qualitative Research Practice: A Guide for Social Science Students and Researchers. In: Richie J, Lewis J, editors. Qualitative Research Practice: A Guide for Social Science Students and Researchers. London: Sage Publications; 2003. p. 219–262.
32. Bojke L, Bogdan G, Jankovic D, Peters J, Soares M, Stein K. Informing Reimbursement Decisions Using Cost-Effectiveness Modelling: A Guide to the Process of Generating Elicited Priors to Capture Model Uncertainties. PharmacoEconomics. 2017;35:867-877.
33. Soares MO, Sharples L, Morton A, Claxton K, Bojke L. Experiences of Structured Elicitation for Model-Based Cost-Effectiveness Analyses. Value in Health. 2018;21(6):715-723.
34. Merkens H. Selection Procedures, Sampling, Case Construction. In: Flick U, von Kardorff E, Steinke I, editors. A Companion to Qualitative Research. London: Sage Publications; 2004. p. 165-71.
35. Iglesias CP, Thompson A, Rogowski WH, Payne K. Reporting guidelines for the use of expert judgement in model-based economic evaluations. Pharmacoeconomics. 2016;34(11):1161–72.

Tables

Table 1: Summary of modeller characteristics (in-depth interviews)

Number of Informants (M1-M24)	**Gender of informant**		**Nature of work**		**Location of work**		**Level of modelling experience**		**Nature of role**		**Number of models worked on**	
	Male	16	Academic	16	UK	17	Senior	9	Supervisory only	8	1-5 models	8
	Female	8	Non-academic	8	Canada	7	Junior	15	'Hands-on' modelling	10	6-15 models	5
									Combination of both	6	16+ models	11

Table 2: Summary of case study characteristics and informants

	Case study context	Number of modellers in immediate structural development	Nature/background of modellers	Number/background of clinicians in immediate structural development	Other (wider) modelling team members
Case Study A (CSA)	Academic, university setting, UK.	Two (M25 and M26).	M25 – Junior modeller, 'hands-on' model development M26 – Senior modeller, supervisory role.	One (C1). Chief Investigator for the clinical project and a specialist consultant with over ten years' experience working for a local NHS hospital. C1 had no previous experience of health economic modelling.	Additional modeller ad-hoc input. Broader team of clinical collaborators and statisticians.
Case Study B (CSB)	Policy institute, UK.	Two (M27 and M28).	M27 – Junior modeller, 'hands-on' model development. M28 – Senior modeller, supervisory role	Up to eleven per meeting (C2 – C12) – clinicians from different geographical locations and clinical roles. Three interviewed (C2, C3, and C4). All worked for NHS hospitals and had over ten years' experience as consultants in respective specialisms. All had some previous exposure to modelling/projects with cost-effectiveness element.	Information specialists and project managers.

Table 3: Summary of results

Summary of results
Clinician recruitment: - Modellers described different processes for recruiting clinicians to structural development - Most UK academic, industry and policy modellers used formal methods: established projects/links - Using informal methods (UK/Canadian academics) made accessing clinicians difficult.
Numbers and backgrounds of clinicians: - Numbers varied according to the context of modellers' work and the nature of the project - Several modellers (n=6) and Case Study A worked on projects with only one clinician - Modellers emphasised the importance of working with senior or "key" clinicians.
Problems with clinician involvement: *Clinician numbers:* - Modellers discussed problems with structural generalisability with only one clinician - In Case Study A, one clinician struggled to decide on representative pathways to inform structure - Diversity of clinician experience was not acknowledged as important by most modeller informants *Clinician understanding:* - Modellers encountered difficulties with retrieving information from clinicians to inform structure - Clinicians did not communicate 'important' events which could impact cost-effectiveness - Clinicians struggled with interpretation of terminology used by modellers during discussions.
Recommendations from the research: - Discussions between multiple clinicians is valuable in developing representative structures - Clinicians should be recruited from diverse backgrounds to enhance structural generalisability - Structural processes would benefit from training clinicians in modelling.

Table 4: Number of clinicians typically involved in structural development (reported by interview informants in phase one)

Number of clinicians worked with on any project	Number of responses from informants
0 clinicians	1
1 clinician	6
2 clinicians	11
3 clinicians	7
4 clinicians	5
5 - 9 clinicians	2
10 - 12 clinicians	6

Appendix 1: Interview topic guide for in-depth interviews with modellers (phase one)

Experience of modelling

- How did you begin?
- What is your current role? What does it involve?
- Have you worked on many models?
- What type of modelling have you done previously? Disease areas? Model types?

General modelling questions

- How would you define the structure of a model? What are the boundaries?
- What would you define as a 'good' modelling outcome?

Examples of modelling activity

- Can you think of an example of a model which you have worked on, where the development process was done particularly well?
- Can you think of any examples of modelling processes which contrast with this?
- Have you found the process easier/harder depending on the particular disease area that you are working in?
- Have you found the process easier/harder depending on the clinician(s) that you speak to? The number of clinicians that you speak to?
- Do you think that there is an ideal way to model?

Model-building guidance

- Do you or have you ever used modelling guidance to assist you in model building? Which guidance? How did you use it?
- What do you think to the published modelling guidance as a whole?

Model building process

- Can you talk me through the process by which you usually develop the structure of a decision-analytic model? Why do you include [x] stage? Are there any other stages that you could include?
- Do you speak to clinicians? How many? At what stage(s)? How do you 'recruit' them?
- What model checking activities are carried out? In terms of the model structure?

- Have you worked in modelling teams where this process is done differently?

Future research

- Which aspects of the model building process do you think require further investigation [for the purpose of developing best practice guidance]?
- Can you think of teams of modellers who would provide interesting case studies for the purpose of this research?

Finish

- Anything else that you would like to contribute?